



Lost Between Services: A Narrative Review of Loss to Follow-Up in the Transition from Paediatric to Adult Congenital Heart Care

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Received: 19 March 2023; **Accepted:** 27 July 2023; **Published:** 18 October 2023

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Abstract

Almost every child born with a heart defect now reaches adulthood, and almost every one of them needs cardiac review for the rest of their life. A sizeable minority stop attending in their late teens, and many are not seen again until something goes wrong.

Objectives: *To review the published evidence on discontinuity of cardiac follow-up around the transfer from paediatric to adult congenital heart care: how many young people are lost, which of them, why, what it costs, and what has been shown to help.*

Material and Methods: *Narrative review of cohort studies, cross-sectional surveys, population registries, randomised trials and society guidance indexed in PubMed and the major cardiology and paediatric journals. No new patient data were generated; every estimate quoted is one published by the original investigators.*

Results: *Pooled across 17 studies and 6847 patients, 26.1 per cent of young people discontinued cardiac follow-up around the transitional age, with individual estimates ranging from 3.6 to 62.7 per cent. Pooled discontinuity was 34.0 per cent in the United States, 25.7 per cent in Canada and 6.5 per cent in Europe. In a survey of 922 adults attending a specialist clinic for the first time, 42 per cent reported a gap of more than three years and 8 per cent a gap of more than a decade, with the first lapse at a mean age of 19.9 years. Gaps were commonest in the mildest disease. A nurse-led transition intervention made a timely first adult appointment 1.8 times more likely at the recommended transfer point and 3.0 times more likely six months later.*

Conclusion: *Loss to follow-up is not random. It concentrates in young people who feel well, who were told their repair was a success, and who were never given a reason to return. The interventions that work are administrative and relational rather than technical, and the wide regional variation suggests how much of the problem is built into how services are organised.*

Abbreviations: *ACHD – Adult Congenital Heart Disease, CHD – Congenital Heart Disease, CI – Confidence Interval, HR – Hazard Ratio, RCT – Randomised Controlled Trial.*

Keywords: *Congenital Heart Disease; Transition to Adult Care; Loss to Follow-Up; Continuity of Care; Adolescent Health*

Introduction

Congenital heart disease used to be a paediatric subject because most of those born with it did not become adults. That has changed within a single professional lifetime. Survival to adulthood rose steadily through the closing decades of the twentieth century [1], mortality fell across all lesion groups [2], and population data from Quebec covering 2000 to 2010 confirmed what the trend implied: adults with congenital heart disease now outnumber children with it [3].

Those adults are not cured. A repaired tetralogy of Fallot, a Fontan circulation, even a closed septal defect leaves a heart that behaves differently over decades [38], and guidelines on both sides of the Atlantic recommend lifelong review at intervals set by the lesion [4,5]. The point at which that review is most likely to break down is the handover from paediatric to adult services, which coincides with leaving school, leaving home and feeling perfectly well [6,7].

This review sets out what is known about that breakdown: how many young people discontinue follow-up, which of them, what reasons they give, what happens to those who return, and which interventions have been tested. The title's phrase is deliberate. These patients are not usually discharged and they do not usually refuse care. They fall into the space between two services, each of which assumes the other is holding them.

Material and Methods

Search strategy: PubMed and the archives of the major cardiology, paediatric and nursing journals were searched for studies of transition, transfer, care gaps and loss to follow-up in congenital heart disease. Search terms combined congenital heart disease and adult congenital heart disease with transition, transfer, loss to follow-up, discontinuity, care gap, retention and adolescent. Reference lists of society statements [4,5,8,9] were screened by hand for primary sources.

Eligibility: Studies were included if they reported the proportion of patients discontinuing or interrupting cardiac follow-up, predictors or stated reasons for doing so, consequences of an interruption, or the effect of an intervention intended to prevent one. Qualitative studies were included where they document reasons given by patients themselves.

Data extraction and handling: Design, setting, sample size, the definition of discontinuity used and the reported estimate were extracted as published. Definitions differ substantially between studies, and the definition is reported alongside every figure because it accounts for much of the spread. No pooling or re-analysis was performed. Figures 1 to 4 and Tables 1 and 2 reproduce published values only. This is a narrative synthesis and was not registered as a systematic review.

Results

How many are lost: The best single estimate comes from a systematic review and meta-analysis of 17 studies enrolling 6847 patients, which found a pooled discontinuity proportion of 26.1 per cent (95% CI 19.2 to 34.6) at the transitional age; under a worst-case assumption about patients whose status was unknown, the pooled estimate rose to 31.9 per cent [10]. Roughly a quarter to a third, then. What that single number conceals is the spread: individual studies reported anywhere from 3.6 to 62.7 per cent (Figure 4).

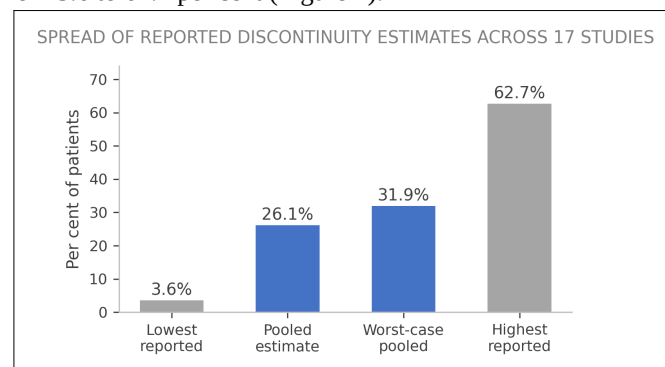


Figure 4: Range of reported discontinuity proportions across the 17 studies in the pooled analysis, against the pooled and worst-case estimates [10]

Where they are lost: Much of that spread is geographical. Pooled discontinuity was 34.0 per cent (95% CI 24.3 to 45.4) in studies from the United States, 25.7 per cent (17.0 to 36.7) in Canada and 6.5 per cent (5.3 to 7.9) in Europe (Figure 1) [10]. A fivefold difference between health systems is not a difference in disease, and the authors point to universal access and to systematically organised transfer as candidate explanations. Within Europe, structural factors have also been implicated: outpatient volume and the staffing available to a unit predicted whether continuity was maintained across transfer [11].

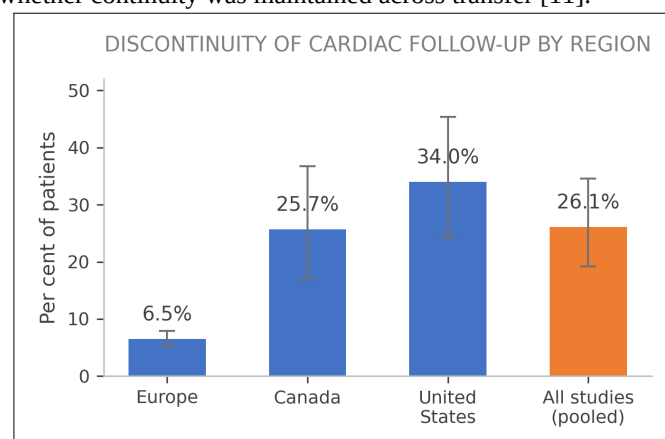


Figure 1: Pooled discontinuity of cardiac follow-up at the transitional age by region, with 95% confidence intervals [10]

Who is lost: The intuitive expectation is that the sickest patients drop out. The opposite is reported. In a survey of 922 adults presenting for the first time to one of 12 specialist clinics, 42 per cent described a gap of more than three years in cardiology care and 8 per cent a gap longer than a decade; the

mean age at the first gap was 19.9 years. Gaps were reported by 59 per cent of those with mild disease, 42 per cent with moderate and 26 per cent with severe disease (Figure 2) [12]. The meta-analysis found the same gradient, with more discontinuity in simple defects (33.7 per cent) than moderate ones (25.7 per cent) [10]. Earlier cohort work pointed the same way, identifying distance from the centre, simpler lesions and the absence of a recent procedure as risk factors [13,14,15].

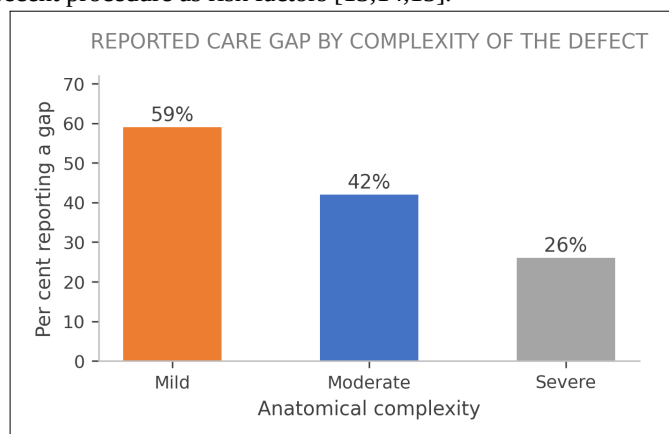


Figure 2: Proportion reporting a gap of more than three years in cardiology care, by anatomical complexity, among 922 adults [12]

Why they are lost: The reasons patients give are unglamorous. The commonest in the survey above were feeling well, not knowing that follow-up was required, and being absent from medical care altogether [12]. Qualitative work with young people in Sweden describes the same thing from the inside: an absence of any explicit handover, uncertainty about who was now responsible, and no felt reason to attend when asymptomatic [16]. A systematic review of transition experiences finds the same themes recurring across studies and countries [33], and the problem was named plainly a decade ago as one of being out of sight and therefore out of mind [37]. Paediatric cardiologists themselves report inconsistent referral practice and uncertainty about where to send patients [17], and transfer practice across centres in Europe and North America has been documented as highly variable [18]. Psychological factors compound this: anxiety and depression are more common in this population than in the general adult population and are frequently unrecognised [19,20,21].

Study	Design and cohort	Principal reported finding
Moons et al. 2021 [10]	Meta-analysis, 17 studies, 6847 patients	Pooled discontinuity 26.1%; range 3.6-62.7%; Europe 6.5% vs USA 34.0%
Gurvitz et al. 2013 [12]	Survey, 922 adults, 12 ACHD centres	42% with gap >3 years; 8% >10 years; first gap at mean age 19.9 years
Reid et al. 2004 [6]	Cohort of young adults with complex defects	Fewer than half had successfully transferred to adult care
Mackie et al. 2009 [7]	Population database, Quebec	Lapses in care common; concentrated around late adolescence
Mackie et al. 2018 [22]	Cluster RCT, 121 adolescents	Nurse-led intervention: HR 1.8 at transfer, 3.0 at six months
Mylotte et al. 2014 [23]	Population database, 71,467 patients	Specialised ACHD care independently associated with lower mortality
Yeung et al. 2008 [24]	Cohort of adults returning after a lapse	Lapse of care predicted subsequent morbidity and need for intervention

Table 1: Principal studies reviewed, with the finding as reported by the original investigators

What it costs: A gap is not merely administrative. Adults returning after an interruption were more likely to require urgent intervention than those who had remained under review [24], and patients unseen for more than five years have been found on re-presentation to have accumulated correctable lesions [25]. Hospitalisation patterns shift sharply upward across the transition years [26,27]. At the population level, the counterfactual is visible: in Quebec, referral to specialised centres rose from 1997 and mortality in this population began to fall three years later, with specialised care independently associated with reduced all-cause mortality across 71,467 patients [23]. Contemporary tertiary-centre data show that death in this group is now predominantly cardiovascular and often occurs at ages where intervention was still possible [28].

What helps: The strongest evidence is for structured, person-delivered transition preparation. In a cluster randomised trial of 121 adolescents aged 16 to 17 with moderate or complex disease, two individualised nurse-led sessions made a first adult appointment within a month 1.8 times more likely at the recommended transfer point (95% CI 1.1 to 2.9) and 3.0 times more likely six months later (1.1 to 8.3), and improved knowledge and self-management scores (Figure 3) [22]. A later trial in younger adolescents improved transition readiness on a validated instrument [29], and a person-centred programme has been evaluated in a hybrid randomised design [30]. Society guidance now sets out programme components explicitly [8,9,31], and a global consensus statement covers transfer as well as transition [32]. Retention deserves as much attention as transfer, since patients can be lost inside a single institution as readily as between two [34], and transition has been argued to require explicit design rather than assumption [36].

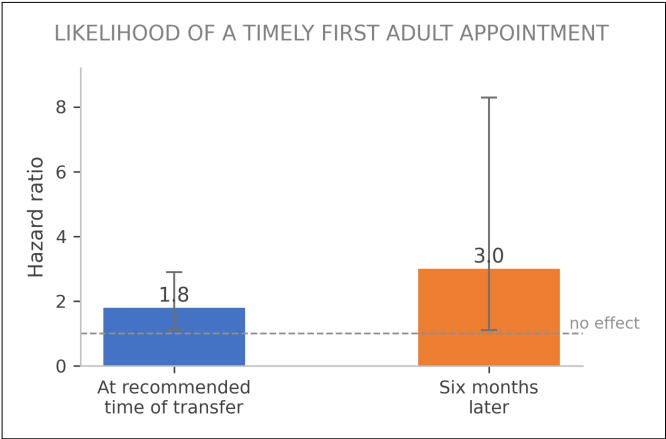


Figure 3: Hazard ratio for attending a first adult appointment within one month, nurse-led intervention versus usual care, with 95% confidence intervals [22]

Factor	Direction of association	Evidence
Mild or simple defect	More likely to lapse	Consistent across survey and pooled data [10,12]
Feeling well / asymptomatic	Commonest stated reason	Patient-reported in survey and qualitative work [12,16]
Not knowing follow-up is needed	Commonest stated reason	Patient-reported; reflects handover failure [12,16]
No explicit transfer process	More likely to lapse	Variable practice documented across centres [17,18]
Lower unit volume and staffing	More likely to lapse	Structural predictor of discontinuity [11]
Structured transition preparation	Less likely to lapse	Randomised evidence [22,29]
Universal access health system	Less likely to lapse	Regional differences in pooled data [10,23]

Table 2: Reported predictors of, and protective factors against, discontinuity of follow-up

Discussion

Three things stand out. The first is that loss to follow-up is patterned, not random. It concentrates in the young people whose disease is mildest, who feel well, and whose childhood care ended on a note of reassurance. The clinical reasoning behind that reassurance is usually sound; the problem is that reassurance about the present is routinely heard as discharge from the future.

The second is that the variation between health systems is larger than the variation between patients. A pooled 6.5 per cent in Europe against 34.0 per cent in the United States is not explained by anatomy or by adolescent behaviour [10]. It points at insurance, at whether a named adult service exists to receive the patient, and at whether anyone is accountable for the handover. That is an encouraging finding, because system design is modifiable in a way that adolescence is not.

The third is that what works is unglamorous. The intervention with randomised support is a nurse having two structured conversations with a seventeen-year-old [22]. It is worth being precise about what that trial showed and did not show: the outcome was the timeliness of the first adult

appointment, in 121 adolescents with moderate or complex disease, at two Canadian centres. It did not demonstrate an effect on long-term retention, on morbidity or on mortality, and the confidence interval at six months is wide. It is the best available evidence, not conclusive evidence.

The link from discontinuity to harm is likewise indirect but consistent. Patients who lapse present later and worse [24,25], hospitalisation rises across these years [26,27], and specialised care is associated with survival at population scale [23]. No trial has randomised young people to continuity or its absence, and none will. The inference has to be drawn from converging observational evidence, and it should be stated as such rather than dressed up.

Study limitations

The limitations belong to the underlying literature. There is no agreed definition of discontinuity: studies variously use a gap of two years, three years, or failure to attend a first adult appointment, which alone accounts for much of the 3.6 to 62.7 per cent spread [10]. Much of the evidence is retrospective, and cross-sectional surveys recruit at specialist clinics, meaning they sample patients who came back and cannot describe those who never did [12]; active case-finding for young people who have simply disappeared from services has only recently been attempted [35]. Qualitative work is small-scale and single-country [16]. Randomised evidence is confined to two trials at a small number of centres with surrogate endpoints [22,29]. Registry analyses capture attendance rather than adequacy of care [3,23]. And publication is concentrated in high-income settings, so almost nothing here describes the countries where most children with congenital heart disease are now surviving to adulthood [32].

Conclusion

Between a quarter and a third of young people with congenital heart disease stop attending cardiac follow-up around the age at which they are meant to move to adult services, and the ones most likely to disappear are the ones whose hearts were described to them as fixed. The consequences surface years later, in a clinic they were not attending, with a lesion that had been progressing quietly. What the evidence supports is neither a new investigation nor a new drug: a named adult service, an explicit handover with a date attached, and someone whose job it is to notice when a young person does not arrive. The fivefold difference in discontinuity between health systems is the clearest indication of how much of this is within the profession's gift to fix.

Acknowledgements

The authors thank the investigators whose cohorts, registries and trials are drawn together in this article, and the young people who contributed to the qualitative work cited here.

Funding

No dedicated funding from public, commercial or charitable sources supported the preparation of this review.

Conflict of Interest

The authors declare no competing interests.

Ethics Statement

This article reviews previously published studies only. No new patient data were collected and no participants were recruited, so ethical approval was not required.

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Citation: Bakker N, Esposito C, Kelly R. "Lost Between Services: A Narrative Review of Loss to Follow-Up in the Transition from Paediatric to Adult Congenital Heart Care." *J Clin Case Stud* (2023): 001-006 doi: 10.5281/zenodo.22227597